

CASE REPORT

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PICA flow-related aneurysms and posterior fossa AVM: rare association and challenging management: case presentation and review of literature

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Abstract

Background The association of an infratentorial arteriovenous malformations (AVMs) with a PICA aneurysm is very rare and the presence of associated aneurysms was an independent predictive factor of poor outcome at follow-up for posterior fossa AVMs (pfAVMs). We report the case of a 57-year-old female patient admitted to our institution with a challenging management of a pfAVM associated with multiple flow-related aneurysms of the PICA.

Case presentation A 57-year-old patient with no medical past history was admitted to our emergency department with a sudden onset headache and severe neck stiffness. A cerebral CT Scan showed a cerebellar hematoma. Digital subtraction angiography demonstrates a right paramedian supero-posterior infratentorial AVM located at the postero-superior part of the right cerebellar hemisphere with a small compact nidus supplied by branches of the right PICA and the right superior cerebellar artery. Venous drainage being toward the torcular. The AVM is associated with 3 flow-related aneurysms. We opted for surgical treatment of the 2 high-flow aneurysms on the distal branch of the right PICA. Patient was then sent for radiosurgery for the AVM. We opted for observation of the left small aneurysm.

Conclusion Microsurgery for PICA aneurysms is particularly well suited because exposure through a far-lateral craniotomy is excellent. Endovascular embolization can be used to occlude the aneurysm as well as treat the AVM at the same time in certain circumstances. Clipping or coiling the aneurysm should be discussed in a multidisciplinary meeting.

Keywords Flow-related aneurysm, Posterior fossa AVM, Case report

Introduction

Posterior inferior cerebellar artery (PICA) aneurysms are the second most common group of aneurysms in the posterior cerebral fossa (PCF). The basilar trunk aneurysms is the first group. Posterior fossa arteriovenous

malformations (pfAVM) are rarer than supratentorial AVMs. These particular AVMs represent 7–15% of brain AVMs [1]. AVMs are defined by the presence of an arteriovenous shunt through a nidus of coiled and tortuous blood vessels that connect feeding arteries to draining veins. They are generally accompanied with flow-related aneurysms [2]. PfAVMs are easily the site of associated aneurysms, specifically feeder vessel aneurysms. Compared with supratentorial AVMs, feeder vessel aneurysms are also more likely to cause posterior fossa hemorrhage [3]. The association of an infratentorial AVMs with a

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PICA aneurysm is very rare and the presence of associated aneurysms was an independent predictive factor of poor outcome at follow-up for pfAVMs [4].

Separately, these 2 vascular malformations represent a therapeutic challenge for vascular neurosurgeons and interventional neuroradiologists. Their association even more. There are 3 therapeutic options including surgery, radiosurgery, and endovascular embolization which are most often all associated. The choice of the best therapeutic strategy for PICA aneurysm associated with AVM still remains to be defined. Several factors must be considered, in particular the vascular angioarchitecture of the AVM and the aneurysm, the arterial segment of the PICA affected, and of course the expertise of each team and the technical platform available.

Despite the development of radiosurgery and endovascular techniques in recent decades, microsurgery remains the gold standard treatment for most of those lesions [1].

We report the case of a 57-year-old female patient admitted to our institution with a challenging management of a pfAVM associated with multiple flow-related aneurysms of the PICA.

The objective of our case presentation is to raise the surgical and endovascular therapeutic difficulties of this association, to make a brief reminder of the surgical anatomy and the clinical importance of the PICA, and a brief review of the literature on the management.

Case presentation

Clinical presentation

A 57-year-old patient with no medical past history was admitted to our emergency department with a sudden onset headache and severe neck stiffness. The Glasgow Coma Scale was 15.

Diagnosis assessment

A cerebral CT Scan showed a cerebellar hematoma with filling of the basal cisterns. Cerebral MRI showed a posterior fossa hematoma with a possible vermian AVM. Digital subtraction angiography (DSA) demonstrates a right paramedian supero-posterior infratentorial AVM located at the posterosuperior part of the right cerebellar hemisphere with a small compact nidus of 15 mm in diameter supplied by branches of the right PICA and the right superior cerebellar artery; venous drainage toward the torcular (Fig. 1).

The AVM is the seat of a small aneurysm (2.94×2.34 mm) in its anterior part, associated with a prenidial dysplastic two flow-related aneurysms at

the level of the right PICA before the bifurcation for the big one (n1) (11.5×2.67 mm); the second one (n2) (4.64×6.93 mm) on the superior branch of PICA bifurcation and a small dysplastic aneurysm (n3) (3×1.7 mm) of the contralateral PICA.

Management

We opted for surgery of the 2 high-flow aneurysms on the distal branch of the right PICA. Under general anesthesia, the patient was operated in a modified Park-bench position, and a right sub-occipital lateral approach was performed (Fig. 2). We performed a lateral sub-occipital craniectomy, we removed the posterior arch of C1 for better exposure. After opening the dura mater, the intradural part of the vertebral artery was identified, followed and dissected gently for the proximal control. Once the telovelotonsillar segment of the right PICA and the aneurysms had been identified, aneurysm n1 was excluded using a fenestrated clip (Fig. 3), and then baby aneurysm n2 was coagulated.

Follow-up

The patient was conscious with GCS at 15 without any deficit after surgery. She was then sent for radiosurgery for the AVM. We opted for observation of the left small aneurysm (n3). Follow-up Imaging at 6 months shows exclusion of the aneurysm clipped and stability of the small one. The AVM has not rebleed and the patient is still awaiting radiosurgery treatment due to financial conditions.

Discussion

We present the case of a 57-year-old female patient, admitted for a subarachnoid hemorrhage WFNS 1 FISHER 4 in whom the DSA showed a Spetzler Martin grade 2 vermian AVM associated with multiple high-flow aneurysms treated surgically. The particularity is the difficulty of the decision-making in such cases.

Anatomy of PICA, clinical consideration and therapeutic implications

The PICA is the VA's principal, most clinically significant branch. The PICA is divided in five segments: anterior medullary, lateral medullary, tonsillomedullary, telovelotonsillar, and cortical segments [5–7].

The clinical functions of the PICA are significant. His anatomy is complex. His tortuous course makes the endovascular treatment of PICA aneurysms difficult, especially when distal. The PICA is also easily exposed during surgery.

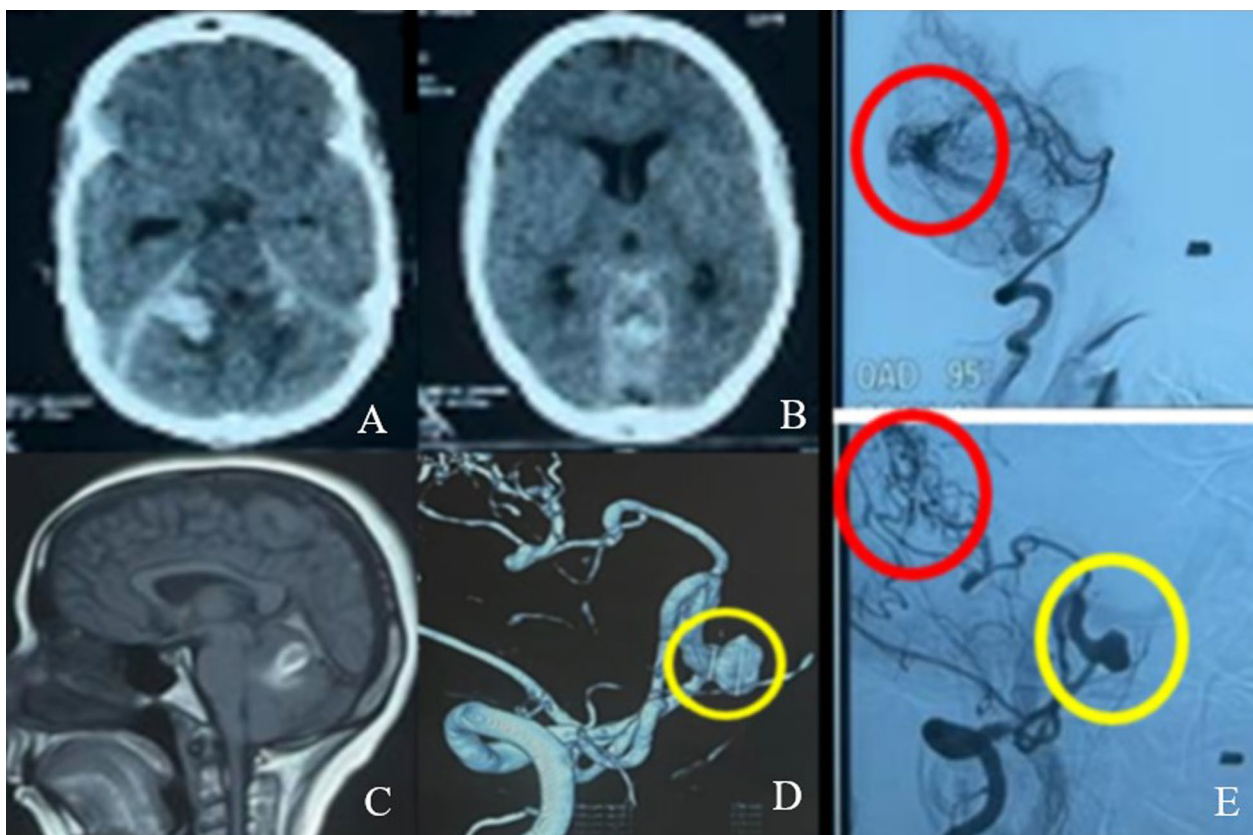


Fig. 1 A–B–C: Cerebral CT and MRI showing a cerebellar hematoma with subarachnoid hemorrhage D–E: DSA shows a posterior fossa AVM feed by the right PICA. Venous drainage towards the torcular and a large flow related PICA aneurysm

In our clinical case, the aneurysms are located at the telovelotonsillar segment of the PICA making them easy to access surgically compared to endovascular treatment in which complete occlusion of the parent vessel is decided and often performed for exclusion of the aneurysm.

Occlusion of the PICA can result in lateral medullary syndrome. Other symptoms of PICA infarction are dysphagia, dysarthria, and hoarseness, ataxia, dizziness, vertigo, nystagmus, and homolateral cerebellar signs, an ipsilateral Horner's syndrome.

However, with a low risk of ischemia, parent artery sacrifice can be helpful and well-tolerated treatment option for distal PICA aneurysms in some cases [8].

Considering the risk of complications after PICA occlusion, surgery must remain a better option for the patient if the aneurysm is easily accessible, as is the case in our patient.

Management

We cannot therefore decide on the most effective or recommended treatment in the management of PICA aneurysms. Once again multidisciplinary discussion and management is crucial to obtain the best cure rate without increasing procedural risks. None of the techniques (microsurgical resection or endovascular treatment) is superior to the other since there is no head-to-head comparison between these 2 approaches. However, it is necessary to privilege the one which ensures a better functional and vital prognosis for the patient [9].

PICA flow-related aneurysms are generally, as in our case, high-flow aneurysms and are subject to an increased risk of life-threatening re-bleeding [10].

It is, therefore, judicious to set up a therapeutic strategy that allows a complete obliteration of both the AVM and the aneurysm without occluding if possible the PICA seeing the complications caused by its occlusion developed above [11]. The clipping-versus-coiling debate continues



Fig. 2 Patient position and incision mark

to haunt neurosurgeons. The tradeoffs between minimally invasive coil embolization versus the more durable clip ligation continue to complicate decision-making in aneurysm surgery. On one hand, the small caliber of the PICA and the broad neck of the associated aneurysms place the PICA at some risk during coil embolization. On the other hand, due to a great number of adjacent structures and the complexity of the anatomy, even with multiple variations existing, the PICA is quite possibly injured in surgery [7, 12–14].

The optimal management of PICA flow-related aneurysms associated with AVMs is still controversial. Some studies demonstrated AVM should be treated first because the associated diseases would cure after the removal of AVM [15, 16]. Other studies suggested the treatment of aneurysms should be prior due to the higher

risk of rupture [17]. In some cases, the PICA aneurysm and AVM could be treated simultaneously, with surgery or endovascular embolization [18]. Each case is different and the attitude will depend not only on the angioarchitecture of the AVM and the aneurysm but also on the technical platform and the expertise of each team.

Stereotaxic Radiosurgery (SRS) remains a therapeutic arsenal of choice in the management of AVMs. It involves the delivery of localized high-dose radiation to the AVM which leads to gradual sclerosis of the blood vessels with eventual obliteration over a period of 2 to 3 years. Successful treatment with SRS depends on AVM size, grade, location, angioarchitecture, density of the nidus, and radiation dosage. AVMs smaller than 3.5 cm are ideal for obliteration. Due to the relatively long time for AVM obliteration, the increased risk of bleeding from

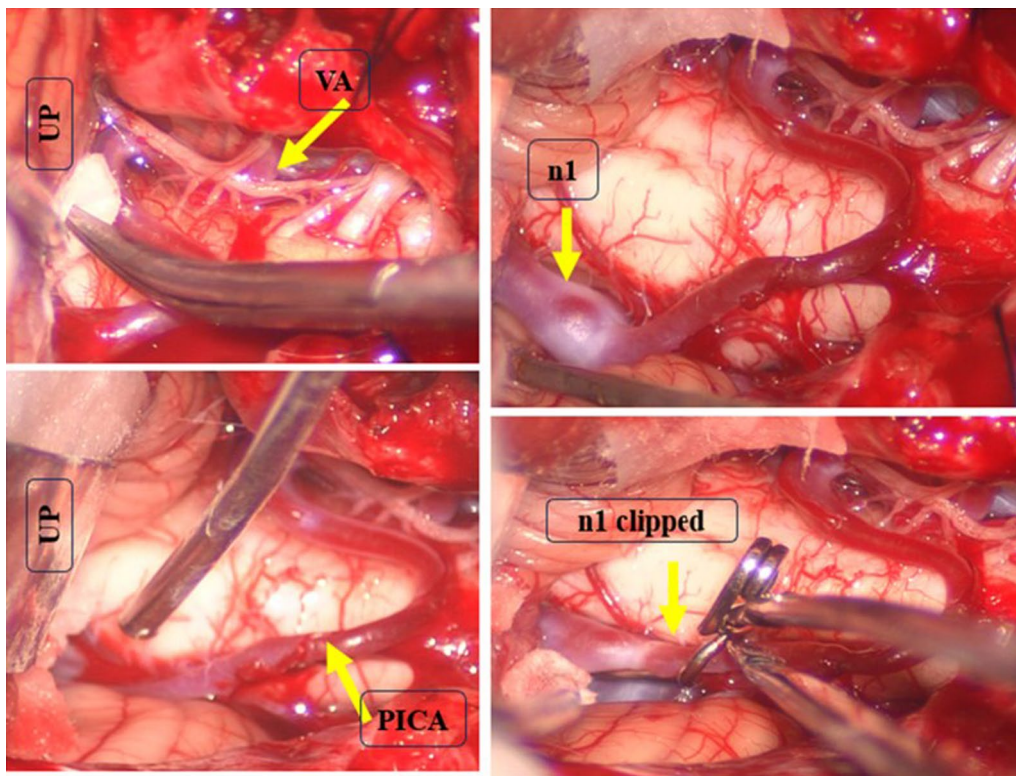


Fig. 3 Per-operative view of the big flow-related aneurysm and its clipping

flow-related aneurysms, these aneurysms should therefore be managed either by clipping or endovascular treatment before starting AVM treatment with radiosurgery [19]. Table 1 summarizes a few articles on AVMs of PCF associated with PICA flow-related aneurysms, their management and the future of patients.

Conclusion

Microsurgical or endovascular management of distal flow-related PICA aneurysms before treatment of the AVMs nidus is recommended to avoid re-bleeding.

Microsurgery for PICA aneurysms is particularly well suited because exposure through a far-lateral craniotomy is excellent, proximal control is immediately accessible, and minimal microdissection is needed to open the tonsillomedullary fissure and follow the PICA and VA. Endovascular embolization can be used to occlude the aneurysm as well as treat the AVM simultaneously in certain circumstances. The clipability versus coilability of the aneurysm should be discussed in a multidisciplinary meeting.

Table 1 Literature review of similar cases compared to our case

Authors	Imaging	Management	Follow-up
Khayat et al. [20]	Partially thrombosed giant right PICA aneurysm -Cerebellar AVM with a nidus < 3 cm supplied by the right PICA	Endovascular treatment of aneurysm Treatment of AVM deferred	No complications 9-month MRI follow-up: decrease in the size of the aneurysm
David Case et al. [9]	Right distal PICA telovelotonsillar segment Aneurysm	Parent vessel sacrifice with Onyx, partial AVM embolization with Onyx	Death
	Aneurysm of distal right PICA in the telovelotonsillar segment + AVM of the superior cerebellar vermis	Parent vessel sacrifice with onyx, AVM treatment deferred	Death
	A small distal left PICA aneurysm in the left telovelotonsillar segment and small AVM in the vermis	Parent vessel sacrifice with NBCA and Onyx, complete AVM embolization with NBCA	No complications
	Right PICA aneurysm with vermian AVM	Parent vessel sacrifice with NBCA, AVM treatment with gamma knife	No complications
Yasuda et al. [21]	Left PICA aneurysm in the telovelotonsillar segment with small AVM in the superior aspect of the vermis	Parent vessel sacrifice with NBCA, AVM treatment deferred	No complications
Al-Jehani et al. [22]	Right telovelotonsillar segment and cerebellar AVM feed by the left superior cerebellar artery	Complete resection of the cerebellar AVM and clipping of the distal PICA aneurysm	No complications
Nonaka et al. [23]	Inferior vermian AVM with a flow-related aneurysm at the left PICA origin	Excision of AVM 24 h after glue embolization of AVM	5 years after AVM resection: regrowth of the aneurysm to 10.2 mm (which was coiled)
Musliman et al. [24]	Two saccular aneurysms on the peripheral left PICA formed an arteriovenous shunt adjacent to the transverse sinus	Embolization of the proximal aneurysm and resection of the arteriovenous shunt lying superficially in the cerebellar hemisphere	Delayed perioperative ischemic deficit
	Left occipital lobe hematoma—Grade II AVM is supplied by the distal branch of the left middle cerebral artery and has drainage into the superior sagittal sinus	Drainage of the hematoma + Left parietooccipital craniotomy + Left lateral sub-occipital craniectomy + Clipping of PICA aneurysm	No complications
Our case	Saccular aneurysm on the left PICA PFC hematoma, mass effect on the fourth ventricle—Upper vermian AVM feed by the right PICA and 2 flow-related aneurysm	Surgical exclusion of the 2 aneurysms on the distal segment of the PICA AVM referred for SRS	No complications at 6 months follow-up

Abbreviations

PICA	Posterior inferior cerebellar artery
AVMs	Arteriovenous malformations
pfAVM	Posterior fossa AVM
PCF	Posterior cerebral fossa
VA	Vertebral artery
CN	Cranial nerve
DSA	Digital subtraction angiography
SRS	Stereotaxic radiosurgery
NBCA	N-butyl 2-cyanoacrylate

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ADKfDP contributed to conceptualization, writing—original draft-editing; SA contributed to writing; BO contributed to writing; BABA contributed to writing, supervision, and validation; EMO contributed to writing; AEO contributed to supervision and validation.

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The Authors certify that they have obtained all appropriate patient consent.

Competing interests

The Authors declare that they have no competing interests.

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